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Case Report

Unicystic Ameloblastoma of Posterior Maxilla- a Rare Entity!

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ABSTRACT

Unicystic ameloblastoma is an uncommon odontogenic lesion, with radiographic and gross appearance suggestive of a jaw cyst. Microscopic features of the lesion, however, reveal classic ameloblastic type lining of the cyst cavity with or without mural nodular growth. Unicystic ameloblastoma typically presents in the posterior mandibular region, in the second decade of life. We report a case of unicystic ameloblastoma in the posterior mandibular region which is a rare site for this lesion. In this case report, 40 years old male presented to the Maxillofacial Department with a history of swelling at the upper 1st premolar region extending to the 3rd molar region. Orthopentogram (OPG) showed a radiolucency and root resorption of the1stand 2nd molar and impacted third molar of the maxilla. This was followed by cone-beam computed tomography (CBCT). The patient was treated by surgical enucleation of the lesion and conjoint extraction of impacted 3rd molar. The microscopic picture was consistent with unicystic ameloblastoma.

Keywords: Unicystic ameloblastoma, impacted 3rd molar, Odontogenic cysts

Introduction

Ameloblastoma is a benign intraosseous tumor. Although rare, it is the commonest odontogenic tumor excluding Odontomas. It accounts for only 1% of all oral tumors. It is a locally aggressive tumor that originates from remnants of the dental lamina and odontogenic epithelium.[1]Approximately 80% are found in mandible while around 14% are seen in the maxilla.[1] The peak incidence of diagnosis is seen in the fourth and fifth decades of life. Radiologically these can be multicystic/nodular and unicystic.[1]According to the World Health Organization (WHO) classification of head and neck tumors types of ameloblastomas include follicular, plexiform, acanthomatous, granular, and basloid[2].

Unicystic ameloblastoma (UA) is defined as ameloblastoma that forms a single unilocular cyst radiologically. Histologically unicystic ameloblastoma typically shows ameloblastic epithelium lining the cyst cavity, with or without mural tumor growth.[3]

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UA is mostly reported in the second and third decades of life. UAs have a slight male predilection and originate from the posterior part of mandible in most cases.

Approximately 10% of UA occurs in maxilla[1]. Approximately 50% of cases are associated with an impacted or unerupted tooth.[3] This report is a rare case of the existence of unicystic ameloblastoma in the maxilla.

Case Report

A 40-year male presents to the Oral and Maxillofacial Department of a tertiary care hospital. The patient had a history of pain in the upper left first premolar region for eight months. There were no co-morbid. The past medical history was not significant, and the patient had no addiction to the pan, "chalia", and smoking. His past dental history revealed a root canal procedure in the upper 1st premolar. The patient later developed painful swelling.

Extra oral examination showed slight facial disfigurement of the left side of the upper face due to swelling, whereas intra oral inspection revealed a fluctuant, diffuse, well-circumscribed painless mass extending medially from maxillary 1st premolar region to distal left side of posterior maxillary region (Figure 1).

It measured approximately 4x3 cm having a smooth shiny surface. There was no bleeding, pus, or any

discharge from the swelling. It was aspirated, fluid was dirty yellow.

On palpation, the mass was soft, fluctuant, well-circumscribed. Orthopentogram (OPG) revealed radiolucency and root resorption of the 1st and 2nd molar and impacted third molar (Figure 2).

Figure 1: Fluctuant, diffuse, well-circumscribed painless mass extending medially from maxillary 1st premolar to left side of the posterior maxillary region

Figure 2: OPG: Unilocular radiolucency extending from 1st premolar to 3rd molar and resorption of 2nd and 3rd molar roots

Cone-beam computed tomography (CBCT) revealed well-circumscribed unilocular radiolucency mesiodistally extending from maxillary left 1st premolar to 2ndmolar, the wisdom tooth is impacted in the radiolucent lesion and is extending from the floor of the maxillary sinus to the alveolar process of 1st premolar to 2nd molar.

Provisional differential diagnosis included dentigerous cyst and odontogenic keratocyst. Excision of the lesion with the removal of impacted tooth was carried out under general anesthesia followed by BIPP (Bismuth Iodine Paraffin Paste) packing to be changed every 15 days for 3 months. The analysis of the surgical specimen revealed a cyst with an impacted third molar. Cyst lining was smooth and tan.

The specimen was transported in formalin and sent for histopathological examination. Histopathological examination revealed multiple pieces of a cyst wall lined by ameloblastic epithelium with loosely arranged cells resembling stellate reticulate. The cyst wall shows cholesterol cleft formation and fibrous tissue (Figure 3&4). The patient had good postoperative progress and is currently under clinical follow-up deprived of signs of recurrence after surgery.

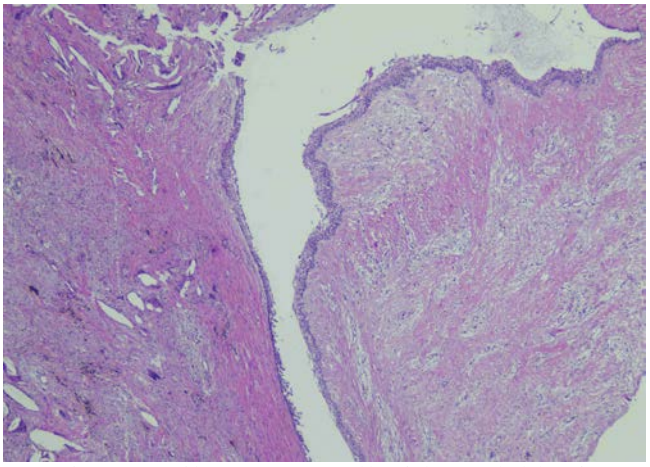

Figure 3: Fibrous cyst wall with ameloblastic epithelial lining. (H&E X 40)

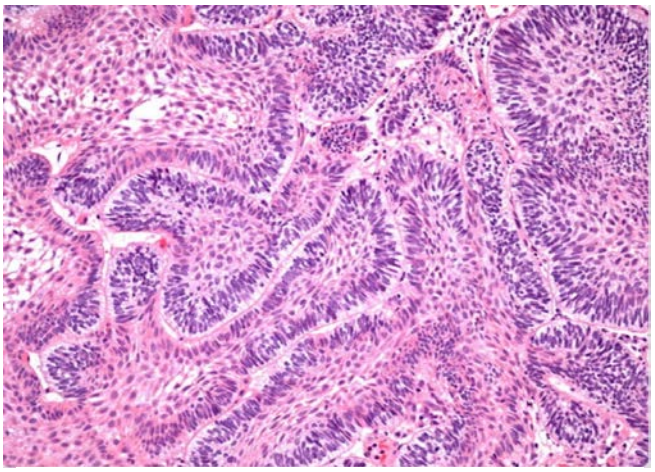

Figure 4: Ameloblastic epithelium showing reverse polarity. (H&E X 100)

Discussion

Unicystic ameloblastoma (UA), a type of ameloblastoma that was first defined by Robinson and Martinez in the last quarter of the 20th century.[4] It refers to those cystic lesions that clinically and radiologically presents as an odontogenic cyst. Microscopic examination, however, demonstrates a characteristic ameloblastic epithelium lining the cyst cavity, with or without mural nodules. The reported frequency of unicystic ameloblastoma is 5-31% of all ameloblastomas. These are slightly more common in male (male to female ratio of 1.5:1) and these are mostly reported in the second and third decade of life.[5]It affects mandible more often than the maxilla. The tumor is mostly observed in the region of the mandibular ramus, while the posterior region of the maxilla is a rare and atypical location.[6]

The tumor is frequently asymptomatic however, large lesions may present as painful swelling of the jaws. The tumor is typically found in association with the crown of mandibular third molar teeth.[7]In our case report, we report it the maxillary region and it was associated with the maxillary impacted third molar tooth. It presented as a painless swelling, facial asymmetry, tooth impaction, tooth displacement, and root resorption.

Radiographically the unilocular lesions present as lytic radiolucency with well-defined sclerotic borders.[8] UA is a less aggressive form of ameloblastomas that can be effectively removed by simple enucleation or other less aggressive surgery.[9] Recurrence rates after conservative treatment of unicystic ameloblastoma are reported to be between 10 and 25%.[5]

The use of Carnoy's solution to decreasing the risk of recurrence after conservative surgical treatment of UA's was initially suggested by Stoelinga and Bronkhorst in 1988.[10] It was suggested that extensive curettage of the bone should be avoided as it may implant foci of ameloblastoma more deeply in the bone.[11] The recurrence rate for UA's after conservative surgical treatment (curettage or enucleation) is generally reported 10–20 %.[12] and on average, <25 %.[13] This is considerably less than 50–90 % recurrence rates which were noted after the conventional curettage of solid or multicystic ameloblastomas.[12, 14] Lau and Samman [15] reported recurrence rates of 3.6 % for resection, 30.5 % for enucleation alone, 16 % for enucleation followed by Carnoy's solution application, and 18 % by marsupialization followed by enucleation, where the lesion is reduced in size.

Conclusion

Herein we report a rare case of unicystic ameloblastoma of the maxilla in a patient of 40 years. Unilocular ameloblastoma shares significant clinical and radiographic similarities with odontogenic cysts and tumors therefore unicystic ameloblastoma should be considered as a possibility in posterior maxillary lesion even in the older age group.

Patient's Consent:

The patient provided both oral and written consent for the information gathering and publication of his case.

CONFLICT OF INTEREST: The author declared no conflict of interest.

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References

1. Radiographic analysis of ameloblastoma: a retrospective study.More C, Tailor M, Patel HJ, Asrani M, Thakkar K, Adalja C.Indian J Dent Res. 2012 Sep-Oct;23(5):698.
2. El Naggar, AK, Chan JKC, Grandis JR. WHO Classification of Head and Neck tumors, 4rth Ed. Lyon, France, IARC, 2017.
3. Li TJ, Kitano M. Reviewing the unicystic ameloblastoma: a clinicopathologically distinct entity. Oral Med Pathol. 1997;2:61–8.
4. Gardner DG. A pathologist's approach to the treatment of ameloblastoma. J Oral Maxillofac Surg. 1984;42: 161–6.
5. Unicystic ameloblastoma: Analysis of 370 cases in a singlecenter in Sri Lanka. Siriwardena BS, Tennakoon TM, Hunter KD. J Oral Pathol Med. 2018;47:706–709.
6. Philipsen HP, Reichard PA. Unicystic ameloblastoma. A review of 193 cases from literature. Oral Oncol. 1998; 34: 317–25.
7. Robinson L, Martinez MG. Unicystic ameloblastoma. A prognostically distinct entity. Cancer. 1977;40:2278-85.
8. Isacsson G, Andersson L, Forsslund H, etal. Diagnosis and treatment planning of unicystic ameloblastoma. Int J Oral Maxillfac Surg. 1986;15:759–64.
9. Handa H, Bailoor DN, Naidu G, Shrivastava K, Raghuvanshi V. Unicystic Ameloblastoma of mandible. Aggressive treatment A myth or a need. Case report and extensive review of literature. IOSR J Dent Med Sci. 2013; 12: 6–31.

10. Stoelinga PJW, Bronkhorst FB. The incidence, multiple presentation and recurrence of aggressive cysts of the jaws. J Craniomaxillofac Surg. 1988;16:184–195
11. Li TJ, Kitano M, Arimura K, Sugihara K. Recurrence of unicystic ameloblastoma: a case report and review of the literature. Arch Pathol Lab Med. 1998;122:371–374.
12. Neville BW, Damm DD, Allen CM, et al. Oral and Maxillofacial Pathology, 3rd ed. St. Louis, Mo: Saunders; 2009.
13. Gardner DG, Corio RL. Plexiform unicystic ameloblastoma. A variant of ameloblastoma with a low-recurrence rate after enucleation. Cancer. 1984;53:1730–1735.
14. Ameerally P, McGurk M, Shaheen O. Atypical ameloblastoma: report of 3 cases and a review of the literatur

Contribution of Authors

- Sana, Sufyan: Substantial contributions to the conception, design of the work; the acquisition, analysis, and interpretation of data for the work
- Sana, Sufyan, Khurram: Drafting the work or revising it critically for important intellectual content
- Sana, Khurram: Final approval of the version to be published
- Sana, Sufyan, Khurram, Ayesha: Agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.